

CASE REPORT

Papillary carcinoma in a thyroglossal duct remnant: A case report and discussion on management



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Abstract Objective: Thyroglossal duct carcinoma is a malignant tumor arising within a thyroglossal remnant. It is a rare entity, seen generally in adults, and characterized by relatively non-aggressive behavior. This case is presented because of its rarity and the presence of cervical lymph node metastasis.

Case report: A 37-year-old female patient presented with a slowly and progressively growing mid-line neck mass. Neck ultrasound and computed tomography scanning revealed a mid-line sub-mental pre-hyoid highly enhancing neck mass, bilateral cervical lymphadenopathy, and tiny nodules in both lobes of the thyroid gland. Fine needle aspiration cytology of the neck mass revealed papillary thyroid carcinoma. The patient underwent total thyroidectomy, Sistrunk procedure, bilateral level I–IV neck dissection, and postoperative I131 ablation and hormonal suppression with thyroxine. The patient has been disease free for 1 year after the operation.

Conclusion: Computed tomography scanning and fine-needle aspiration cytology enhance the preoperative diagnosis. Sistrunk procedure is the standard treatment with a clinically and radiologically normal thyroid gland, while the more aggressive treatment is necessary in advanced cases. The concept of prognostic risk groups should be used to identify patients who would need a more aggressive approach. The prognosis is generally excellent with adequate treatment.

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1. Introduction

A thyroglossal duct cyst (TDC) is the most common anomaly in the development of the thyroid gland. This is the most common form of congenital malformations in the neck as 70% of mid-line masses diagnosed during childhood and 7% in adults are TDCs. It can be located in an area from the base of the tongue to the pyramid of Lalouette and is present characteristically as a mid-line cervical mass at the level of the thyrohyoid

membrane.¹⁻³ Thyroglossal duct carcinoma (TDCa) is a malignant tumor arising within a thyroglossal remnant or a thyroglossal duct cyst. This is a rare condition as 1% of TDCs can harbor malignancy.⁴ A review of the literature showed that 260 cases of malignant thyroglossal cysts have been reported since the first description by Bretano in 1911. The majority of the cases were discovered during pathological examination of TDC specimens.^{3,5}

In this article, we present a case of papillary thyroid carcinoma of a thyroglossal duct remnant because of its rarity, unusual manifestation, and the lymphatic spread, and discuss the current opinions concerning the management.

2. Case presentation

A 37-year-old female patient presented with mid-line neck mass that grew slowly and progressively over 2 years. The mass was intermittently painful. The patient had no history of hoarseness, breathing difficulty or dysphagia. She reported no history of radiation exposure. Physical examination revealed a hard and well circumscribed mass that was mobile with deglutition and protrusion of the tongue. It was fixed to the deep planes of the neck. However, it was not attached to the skin.

Thyroid function tests [serum-free thyroxine (T4), free triiodothyronine (T3), and thyroid-stimulating hormone (TSH)] were within normal limits. Neck ultrasound revealed an echogenic mass of 3.7×2.6 cm in size in the sub-mental region in the mid-line with no cystic changes and bilateral cervical lymphadenopathy, predominantly at level II and III, and more on the right side. The largest lymph node was on the right side and measured 1.3×0.6 cm in size. The thyroid gland was of normal size, shape, and position and few tiny nodules less than 5 mm were seen in both lobes. A computed tomography (CT) scanning of the soft tissue of the neck with and without contrast revealed a mid-line sub-mental pre-hyoid neck mass of 4×2.5 cm in size, highly enhancing, deep to the platysma muscle, and attached to the infra-hyoid muscles (Figs. 1–3). Some heterogeneity was seen in the thyroid gland, while there was no significant lymph node enlargement. Fine needle aspiration cytology of the neck mass revealed papillary thyroid carcinoma.

After multidisciplinary discussion, a fully informed written consent was obtained from the patient, and she underwent total thyroidectomy, Sistrunk procedure, and bilateral level I–IV neck dissection. The strap muscles of the neck were only pushed by the sub-mental mass and were not involved by it. Therefore, they were completely dissected of the sub-mental mass and preserved.

The postoperative histopathology report revealed $3.5 \times 2.6 \times 3$ cm papillary thyroid carcinoma of the thyroglossal duct remnant with negative margins and uninvolved hyoid bone and another focus of papillary micro-carcinoma (0.5 cm) confined to the left lobe of the thyroid gland. Non-keratinizing squamous epithelium and normal thyroid follicles were identified and a focus of vascular invasion was seen in the main Sistrunk specimen. The tumor was partially encapsulated and no invasion into the muscles was seen.

Metastatic papillary thyroid carcinoma was identified in 12 out of 103 lymph nodes within the levels left IIA and III and right IIA and III. The largest metastatic lymph node was at the level right III, and was 13 mm in size.

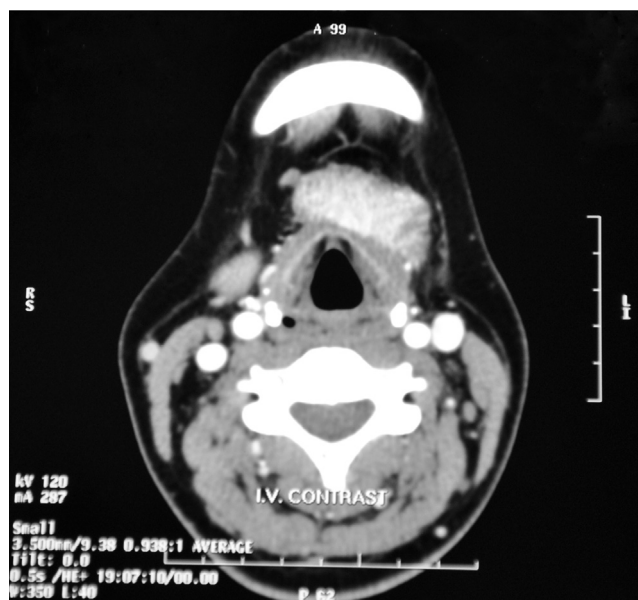


Figure 1 (Axial view) Preoperative post contrast computed tomography scanning of the soft tissue of the neck showing a mid-line pre-hyoid neck mass, highly enhancing, deep to the platysma muscle, and attached to the infra-hyoid muscles.



Figure 2 (Axial view) Preoperative post contrast computed tomography scanning of the soft tissue of the neck showing a mid-line neck mass, highly enhancing, deep to the platysma muscle, and attached to the infra-hyoid muscles.

Postoperatively, the patient had good wound healing, normal voice and breathing, transient hypocalcemia, and uneventful recovery. One month postoperatively, the patient received I131 (120 mCi) followed by hormonal suppression with thyroxine. Ten months postoperatively, I131 whole-body scanning showed no active iodine-avid tissue in the neck region, and revealed no metastasis. Neck ultrasound revealed no residual or recurrent mass. Thyroxine withdrawal stimulated thyroglobulin level was 0.4 ng/ml and thyroglobulin antibody level was negative about 45.5 U/ml. The patient has been a disease free for 1 year after the operation. The Institutional Reviewer

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